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Malignant transformation and tumour recurrence in sacrococcygeal teratoma, a congenital disorder in newborns

Submission date
10/04/2024

Recruitment status
No longer recruiting

Registration date
28/04/2024

Overall study status
Completed

Last Edited
28/11/2024

Condition category
Cancer

[_] Prospectively registered
[X] Protocol
[_] Statistical analysis plan
[X] Results
[_] Individual participant data

Plain English summary of protocol

Background and study aims
Sacrococcygeal teratoma (SCT) is a rare congenital disorder with an estimated incidence of 1: 15,000 to 1:30,000. The treatment is complete resection, but in a substantial proportion of patients, the tumour recurs, often as malignancy. There is scant data on the true recurrence rate of SCT, the timing of recurrence, how recurrence is diagnosed and risk factors for recurrence including histology of the primary tumour. This study aims to assess the SCT recurrence rate, the time after which SCT recurrence occurs, pathology, whether recurrence is a primary or recurrent tumour, and how recurrent SCT is diagnosed.

Who can participate?
All patients with SCT treated by each participating centre during a set period of time will be included. Patients with Currarino triad-associated SCT (arising from defects that occur during embryonic development) are specifically included.

What does the study involve?
Participating centers confirm their participation, the inclusion of consecutive patients in a set period of time and sign a data transfer agreement form which defines data ownership, data use, protection and storage of data and authorship.

What are the possible benefits and risks of participating?
There is no benefit for the individual included patient in this study. However, The United Nations encourages nations to create networks of experts for rare diseases and to increase research support, by strengthening international collaboration and coordination of research efforts and the sharing of data, while respecting its protection and privacy. This collaborative study enables the identification of risk factors for recurrent SCT and essential information regarding malignant SCT transformation. With large, shared patient data sets, it is possible to answer important clinical questions that cannot be answered otherwise to improve the quality of care in every part of the world.

Due to the design of the study including only retrospective patient data, there are no risks for individual patients.

Where is the study run from?
Amsterdam University Medical Centre in the Netherlands

When is the study starting and how long is it expected to run for?
December 2020 to January 2022

Who is funding the study?
KiKa Children's Cancer Free Foundation

Who is the main contact?
Prof. Dr. L.W.E. van Heurn, e.vanheurn@amsterdamumc.nl

Contact information

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Additional identifiers

Clinical Trials Information System (CTIS)
Nil known

ClinicalTrials.gov (NCT)
Nil known

Protocol serial number
KiKa project 440

Study information

Scientific Title
Malignant transformation and tumour recurrence in patients with SacroCoccygeal Teratoma: a global, retrospective cohort study

Acronym
SCT-study

Study objectives
Sacrococcygeal teratoma (SCT) is a rare congenital disorder with an estimated incidence of 1: 15,000 to 1:30,000. The treatment is complete resection, but in a substantial proportion of patients, the tumour recurs, often as malignancy. There is limited data on the true recurrence rate of SCT, the timing of recurrence, how recurrence is diagnosed and risk factors for recurrence including histology of the primary tumour.

Ethics approval required
Ethics approval not required

Ethics approval(s)
The Medical Ethical Board of Amsterdam University Medical Centre (Amsterdam UMC), determined that the Medical Research Involving Human Subject Act (WMO) does not apply to the study and that official approval of the committee was not required (reference number W19_329 # 19.388).

Study design
Multicentre observational retrospective cohort study

Primary study design
Observational

Study type(s)
Diagnostic, Treatment

Health condition(s) or problem(s) studied
Sacrococcygeal teratoma

Interventions
This observational study looks at tumour histology at initial resection and the time between birth and surgery. Furthermore, risk factors associated with recurrence will be analysed.

Collected data included generic and condition-specific variables. Generic variables included: country, gender (male/female/unknown), age at diagnosis (days), pre-operative imaging modalities (none/ultrasound/computed tomography/magnetic resonance imaging/unknown), initial tumour resection at the participating centre (yes/no/unknown), age at initial resection (days), outcome (survival/deceased/unknown), age at follow-up (days), age at death (days), and cause of death. Condition-specific variables were Altman classification (I/II/III/IV/unknown), CS (yes/no/unknown), initial SCT treatment (chemotherapy/surgery/no treatment/unknown), pathology (mature/immature/malignant/unknown), recurrence (yes/no/unknown), the period between birth and recurrence (days), detection of recurrence (clinical examination/imaging/AFP /unknown), serum AFP-level at recurrence (µg/L), recurrent SCT pathology (mature/immature /malignant/unknown) and treatment of recurrent SCT (chemotherapy/surgery/no treatment /unknown). These data were used to calculate the risk of malignant transformation and risk factors associated with sacrococcygeal teratoma recurrence.

Intervention Type
Procedure/Surgery

Primary outcome(s)
The following primary outcomes are measured using patient medical records at a one-time point:
1. Malignant transformation of the initial tumour confirmed by pathology.
2. Risk factors associated with sacrococcygeal teratoma recurrence defined as relapse of the tumour at least three months after initial resection.

Key secondary outcome(s))
The following secondary outcomes are measured using patient medical records at a one-time point:
1. Treatment modality for sacrococcygeal teratoma patients from different income countries
2. Outcomes of treatment for sacrococcygeal teratoma patients from different income countries

Completion date
01/01/2022

Eligibility

Key inclusion criteria
All patients with SCT treated by each participating centre during a set time

Participant type(s)
Patient

Healthy volunteers allowed
No

Age group
All

Sex
All

Total final enrolment
3612

Key exclusion criteria
Patients treated for sacrococcygeal teratoma before 1980

Date of first enrolment
01/12/2020

Date of final enrolment
01/01/2022

Locations

Countries of recruitment
United Kingdom

Afghanistan

Argentina

Austria

Bangladesh

Belarus

Belgium

Brazil

Bulgaria

Cameroon

Canada

Chile

China

Croatia

Czech Republic

Côte d'Ivoire

Denmark

Egypt

Estonia

Ethiopia

Finland

France

Germany

Ghana

Greece

Hong Kong

Hungary

India

Indonesia

Iran

Iraq

Ireland

Israel

Italy

Japan

Korea, South

Latvia

Lithuania

Malaysia

Mexico

Montenegro

Morocco

Netherlands

Nigeria

North Macedonia

Norway

Pakistan

Philippines

Poland

Portugal

Romania

Russian Federation

Serbia

Singapore

Slovakia

Slovenia

Spain

Sri Lanka

Sweden

Syria

Taiwan

Thailand

Tunisia

Türkiye

Ukraine

United States of America

Zambia

Study participating centre
Hospital Garrahan Buenos Aires
Argentina
-

Study participating centre
Hospital Italiano de Buenos Aires
Argentina
-

Study participating centre
Medical University of Graz
Austria
-

Study participating centre
Bangladesh Shishu Hospital & Institute
Bangladesh
-

Study participating centre
Center of Pediatric Surgery of Belarusb
Belarus
-

Study participating centre
Universitair Ziekenhuis Brussel
Belgium
-

Study participating centre
Hôpital des Enfants Reine Fabiola, Université Libre de Bruxelles
Belgium
-

Study participating centre
Federal University of São Paulo
Brazil
-

Study participating centre
University Hospital "St. George"
Bulgaria
-

Study participating centre
UMHATEM "N. I. Pirogov" Sofia
Bulgaria
-

Study participating centre
Yaounde Gynaeco-Obstetric and Pediatric Hospital-faculty of medecine
Cameroon
-

Study participating centre
McGill University, Montreal Children’s Hospital
Canada
-

Study participating centre
Shriners Hospital for Children
Canada
-

Study participating centre
The Hospital for Sick Children
Canada
-

Study participating centre
Hospital de Niños Dr. Roberto del Rio
Chile
-

Study participating centre
Guangzhou Women and Children’s Medical Center
China
510623

Study participating centre
University Hospital centre Zagreb
Croatia
-

Study participating centre
University Hospital of Split and Department of Surgery, University of Split
Croatia
-

Study participating centre
Charles University and University Hospital Motol
Czech Republic
150 06

Study participating centre
Rigshospitalet Copenhagen University Hospital
Denmark
-

Study participating centre
Ain Shams University
Egypt
-

Study participating centre

Kasr Al Ainy Faculty of Medicine
Egypt
-

Study participating centre
Pediatric Surgery Cairo University
Egypt
-

Study participating centre
Tallinn Children's Hospital
Estonia
-

Study participating centre
St.Peter Specialized Hospital
Ethiopia
-

Study participating centre
New Children´s Hospital
Finland
-

Study participating centre
Hôpital des Enfants de Toulouse
France
-

Study participating centre
CHU Rennes, Univ Rennes
France
-

Study participating centre

Limoges University Hospital Center
France
-

Study participating centre
University Hospital Angers
Angers
France
-

Study participating centre
Hospital for sick children La Timone, Assistance publique des hôpitaux de Marseille
Marseille
France
-

Study participating centre
orbonne Université, Armand Trousseau Hopsital –Assistance Publique Hôpitaux de Paris,
Paris
France
-

Study participating centre
Hopitaux Pédiatriques de Nice CHU-Lenval
Nice
France
-

Study participating centre
University Hospital, Poitiers
Poitiers
France
-

Study participating centre
Hôpital Necker-Enfants Malades - APHP GH Centre and Université de Paris Cité
Paris
France
-

Study participating centre
University Hospital of Lille Jeanne de Flandre
Lille
France
-

Study participating centre
Dr. von Hauner Children's Hospital, University Hospital
Munich
Germany
-

Study participating centre
University Medical Center Mannheim, Heidelberg University
Mannheim
Germany
-

Study participating centre
University of Leipzig
Leipzig
Germany
-

Study participating centre
Charité Universitätsmedizin Berlin, corporate member of Freie Universität Berlin and Humboldt Universität zu Berlin
Berlin
Germany
-

Study participating centre
University Hospital Frankfurt/M.
Frankfurt
Germany
-

Study participating centre
Accra College of Medicine
Accra
Ghana
-

Study participating centre
P. & A. Kyriakou Children's Hospital
Athens
Greece
-

Study participating centre
Semmelweis University
Budapest
Hungary
-

Study participating centre
Medical School, University of Pécs
Pécs
Hungary
-

Study participating centre
Li Ka Shing Faculty of Medicine, University of Hong Kong
Hong Kong
-

Study participating centre
Amrita Institute Of medical sciences
Kerala
India
-

Study participating centre
All India Institute of Medical Sciences
Jodhpur

India
-

Study participating centre
All India Institute of Medical Sciences
New Dehli
India
-

Study participating centre
Faculty of Medicine, Public Health and Nursing, Universitas Gadjah Mada, Dr. Sardjito Hospital
Yogyakarta
Indonesia
-

Study participating centre
Research Institute for Children’s Health, Shahid Beheshti University of Medical Sciences
Teheran
Iran
-

Study participating centre
Alkhansaa teaching hospital
Mosul
Iraq
-

Study participating centre
Children's Health Ireland at Crumlin
Dublin
Ireland
-

Study participating centre
Tel Aviv Sourasky Medical Center
Tel Aviv
Israel
-

Study participating centre
IRCCS Istituto Giannina Gaslini
Genoa
Italy
-

Study participating centre
Children's Hospital Bambino Gesù-Research Institute
Rome
Italy
-

Study participating centre
University of Padua
Padua
Italy
-

Study participating centre
Regina Margherita Children’s Hospital
Turin
Italy
-

Study participating centre
Bambino Gesu' Pediatric Hospital
Rome
Italy
-

Study participating centre
Sant'Orsola Hospital, IRCSS, University of Bologna
Bologna
Italy
-

Study participating centre

Meyer Children's Hospital IRCCS Florence
Florence
Italy
-

Study participating centre
Fondazione IRCCS Policlinico San Matteo
Pavia
Italy
-

Study participating centre
Cocody Teaching Hospital at Abidjan
Abidjan
Côte d'Ivoire
-

Study participating centre
Kyoto Prefectural University of Medicine
Kyoto
Japan
-

Study participating centre
Saitama Children's Medical Center
Saitama
Japan
-

Study participating centre
National Center for Child Health and Development
Tokyo
Japan
-

Study participating centre

Graduate School of Medical Sciences, Kyushu University
Fukuoka
Japan
-

Study participating centre
Osaka City General Hospital
Osaka
Japan
-

Study participating centre
Graduate School of Medical Sciences, Kyushu University,
Fukuoka
Japan
-

Study participating centre
Tokyo Metropolitan Children's Medical Center
Tokyo
Japan
-

Study participating centre
Riga Stradins University & Children's Clinical University Hospital
Riga
Latvia
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Study participating centre
Vaikų chirurgijos klinika, Lietuvos sveikatos mokslų universiteto ligoninė Kauno klinikos
Kaunsas
Lithuania
LT-50161

Study participating centre

University Clinic for Pediatric Surgery, Faculty of Medicine, Ss. Cyril and Methodius
Skopje
North Macedonia
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Study participating centre
Faculty of Medicine, Universiti Kebangsaan Malaysia
Kuala Lumpur
Malaysia
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Study participating centre
Hospital Tunku Azizah, Kuala Lumpur Women's and Children's Hospital
Kuala Lumpur
Malaysia
-

Study participating centre
Instituto Nacional de Pediatría
Mexico City
Mexico
-

Study participating centre
Institute for children's diseases, Clinical Centre of Montenegro
Podgorica
Montenegro
-

Study participating centre
University hospital Mohamed VI, Cadi Ayyad University,
Marrakesh
Morocco
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Study participating centre

University Medical Center Groningen
Groningen
Netherlands
-

Study participating centre
Radboud University Medical Center, Amalia Children's Hospital
Nijmegen
Netherlands
-

Study participating centre
Emma Children’s Hospital, Amsterdam UMC, location University of Amsterdam,
Amsterdam
Netherlands
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Study participating centre
University Medical Centre Maastricht
Maastricht
Netherlands
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Study participating centre
Erasmus MC Sophia Children’s Hospital
Rotterdam
Netherlands
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Study participating centre
Amsterdam Public Health Research Institute, Amsterdam UMC, Vrije Universiteit Amsterdam
Amsterdam
Netherlands
1081 HV

Study participating centre

University of Utrecht, Wilhelmina Children's Hospital, UMC Utrecht
Utrecht
Netherlands
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Study participating centre
Lagos University Teaching Hospital
Lagos
Nigeria
-

Study participating centre
Jos University Teaching Hospital
Jos
Nigeria
PMB 2076

Study participating centre
Nnamdi Azikiwe University Teaching Hospital
Nnewi
Nigeria
-

Study participating centre
Abubakar Tafawa Balewa University Teaching Hospital
Bauchi
Nigeria
-

Study participating centre
Oslo University Hospital
Oslo
Norway
4950

Study participating centre

Liaquat National Hospital and Aga khan university
Karachi
Pakistan
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Study participating centre
Children Hospital, Shaheed Zulfiqar Ali Bhutto Medical University
Islamabad
Pakistan
-

Study participating centre
University of Child Health Sciences
Lahore
Pakistan
-

Study participating centre
Philippine Children's Medical Center
Quezon City
Philippines
-

Study participating centre
Philippine General Hospital
Manila
Philippines
-

Study participating centre
The Children's Memorial Health Institute
Warshaw
Poland
-

Study participating centre

Medical University of Gdansk
Gdansk
Poland
-

Study participating centre
Hospital Dona Estefania
Lisboa
Portugal
-

Study participating centre
Hospital Dona Estefânia
Lisboa
Portugal
-

Study participating centre
"Victor Babes" University of Medicine and Pharmacy Timisoara
Timisoara
Romania
-

Study participating centre
FSAI "NMRC of Children Health" MHRF
Moscow
Russian Federation
-

Study participating centre
Volgograd State Medical University
Volgograd
Russian Federation
-

Study participating centre

Irkutsk Regional Children’s Hospital
Irkutsk
Russian Federation
-

Study participating centre
Stavropol State Medical University
Stavropol
Russian Federation
-

Study participating centre
Institute for Mother and Child Healthcare of Serbia "Dr Vukan Cupic"
Belgrade
Serbia
-

Study participating centre
University Children’s Hospital, Center for Pediatric Surgery and Medical Faculty University of Belgrade
Belgrade
Serbia
-

Study participating centre
KK Women’s and Children’s Hospital
Singapore
-

Study participating centre
National Institute of Children´s Diseases
Bratislava
Slovakia
-

Study participating centre

University Medical centre Ljubljana
Ljubljana
Slovenia
-

Study participating centre
Seoul National University Hospital
Seoul
Korea, South
03080

Study participating centre
Asan Medical Center
Seoul
Korea, South
-

Study participating centre
Inje University Busan Paik hospital
Busan
Korea, South
-

Study participating centre
University Hospital Vall d´Hebron
Barcelona
Spain
-

Study participating centre
Children's Hospital La Paz
La Paz
Spain
-

Study participating centre

Virgen del Rocio Children's Hospital
Sevilla
Spain
-

Study participating centre
Hospital Universitario Reina Sofía
Córdoba
Spain
-

Study participating centre
Virgen de la Arrixaca University Clinical Hospital
Murcia
Spain
-

Study participating centre
La Fe University and Polytechnic Hospital
Valencia
Spain
-

Study participating centre
Lady Ridgeway Hospital
Colombo
Sri Lanka
-

Study participating centre
Skane University Hospital Lund
Lund
Sweden
-

Study participating centre

Karolinska University Hospital
Stockholm
Sweden
-

Study participating centre
Astrid Lindgren´s Childrens Hospital
Stockholm
Sweden
-

Study participating centre
Children Hospital
Damascus
Syria
-

Study participating centre
Chang Gung University College of Medicine
Taoyuan
Taiwan
-

Study participating centre
Queen Sirikit National Institute of Child Health
Bangkok
Thailand
-

Study participating centre
Hedi Chaker Hospital University of medecine of Sfax
Sfax
Tunisia
-

Study participating centre

Fattouma Bourguiba Hospital
Monastir
Tunisia
-

Study participating centre
Ege University
Izmir
Türkiye
-

Study participating centre
Istanbul Medeniyet University Faculty of Medicine)
Istanbul
Türkiye
-

Study participating centre
Dr Sami Ulus Maternity and Children Health and Research Application Center
Ankara
Türkiye
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Study participating centre
Faculty of Medicine & Cancer Institute Ankara
Ankara
Türkiye
-

Study participating centre
Hacettepe University, Faculty of Medicine
Ankara
Türkiye
-

Study participating centre

National Specialized Children Hospital “Ohmatdyt“
Kyiv
Ukraine
-

Study participating centre
Leeds Teaching Hospitals
Leeds
United Kingdom
-

Study participating centre
Cambridge University Hospitals NHS Foundation Trust
Cambridge
United Kingdom
-

Study participating centre
Great North Children's Hospital
Newcastle
United Kingdom
-

Study participating centre
Birmingham Children hospital UK
Birmingham
United Kingdom
-

Study participating centre
Royal Manchester Children's Hospital
Manchester
United Kingdom
-

Study participating centre

The Royal London Hospital
London
United Kingdom
-

Study participating centre
Bristol Royal Children's Hospital
Bristol
United Kingdom
-

Study participating centre
UCL Great Ormond Street Institute of Child Health
London
United Kingdom
-

Study participating centre
Faculty of Medicine, University of Southampton
Southampton
United Kingdom
-

Study participating centre
Institute Of Life Course And Medical Sciences
Liverpool
United Kingdom
-

Study participating centre
Royal Hospital for Sick Children
Edinburgh
United Kingdom
-

Study participating centre

Nationwide Children’s Hospital
Columbus
United States of America
-

Study participating centre
University of Utah School of Medicine
Salt Lake City
United States of America
-

Study participating centre
University of Tennessee Health Science Center
Tennessee
United States of America
-

Study participating centre
University of Chicago
Chicago
United States of America
-

Study participating centre
Children’s Mercy Kansas City
Kansas City
United States of America
-

Study participating centre
University Teaching Hospital of Lusaka
Lusaka
United States of America
-

Sponsor information

Organisation
Foundation KiKa

ROR
https://ror.org/05gxzef39

Funder(s)

Funder type
Other

Funder Name
Investigator initiated and funded

Results and Publications

Individual participant data (IPD) sharing plan
Following publication of the study results, the full, anonymous de-identified patient dataset will be made available. Proposals should be directed to sct-study@amsterdamumc.nl to gain access. Data requestors will need to sign a data access agreement.

IPD sharing plan summary
Available on request

Study outputs

Output type	Details	Date created	Date added	Peer reviewed?	Patient-facing?
Results article		06/09/2024	28/11/2024	Yes	No
Participant information sheet	Participant information sheet	11/11/2025	11/11/2025	No	Yes
Protocol file			17/04/2024	No	No